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A true aneurysm of the superficial temporal artery: Is there an underlying pre-disposition to such rarities?

Jake Sloane*, Abdul Aziz, Khalid Makhdoomi

Vascular Surgery Department, Kings Mill Hospital, Mansfield Road, Sutton in Ashfield, Nottinghamshire NG17 4JL, United Kingdom

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ABSTRACT

INTRODUCTION: A 32 year old man presented to our vascular clinic with a lump over the left eyebrow. It had become larger in size and was cosmetically unacceptable.
PRESENTATION OF CASE: Clinical examination of the lump demonstrated a pulsatile swelling consistent with an aneurysm of the superficial temporal artery (STA). Doppler ultrasound demonstrated arterial flow within the lump that could be controlled with pressure over the proximal branch of the vessel.

The STA aneurysm was excised under local anaesthesia by ligation of the feeding and draining branches. It measured 3.4 cm × 3.7 cm. Histological examination confirmed a rare finding of a true aneurysm of the STA.
DISCUSSION: Aneurysms are classified into false or true types, with false aneurysms of the STA accounting for 95% of cases reported. It has therefore been suggested that true aneurysms of the STA may develop from a yet unknown pre-existing vessel condition.

The histological findings in our case demonstrated myxoid/mucoid deposits replacing elastin fibres of the media layer, in addition to cystic lesions seen in the adventitia. These features were thought to be nonspecific but have been reported in rare conditions known as cystic adventitial disease and cystic medial necrosis. Such is their obscurity that we speculate that both of these may represent variations of the same underlying pathology. As myxoid/mucoid changes are also noted in more well-known connective tissue disorders including Marfan's syndrome, imaging was arranged to screen our patient for thoracic and abdominal aneurysms. These proved negative and there were no Marfanoid features noted prior.
CONCLUSION: Aneurysms of the STA are easily treated by surgical excision. This should be considered when they become large, painful or are cosmetically undesirable. Unusual histological findings in the specimen may indicate a connective tissue disorder that should be investigated if indicated.

1. Introduction and presentation of the case

A 32 year old man presented to our clinic with a slowly growing and increasingly painful swelling over the left eyebrow. The swelling had also become cosmetically unacceptable to the patient. Clinical examination demonstrated a pulsatile lump just superior to the lateral aspect of the left eye brow measuring 3.4 cm × 3.7 cm. (Fig. 1) No facial nerve paresis was detected.

Using a handheld Doppler probe, a triphasic arterial signal was demonstrated. Compression of the proximal artery controlled the pulsations and an STA aneurysm was clinically diagnosed. In view of the risk of further expansion and potential rupture, the patient was listed for day case surgery. Under local anaesthesia, the proximal and distal branches to the aneurysm were identified, ligated and the aneurysm excised fully (Fig. 2)

Histological examination of the specimen demonstrated several unusual features. A rare case of a true aneurysm of the STA layers was demonstrated, involving all three layers of the vessel wall. Disruption of the elastin fibres of the wall was seen, being replaced with mucin and myxoid deposits. Several small cystic changes were also noted within the adventitial wall of the vessel, a feature characteristic of another rarely seen vascular pathology, cystic adventitial disease (Fig. 3).

2. Discussion

STA aneurysms are rare and are most commonly associated with head trauma. They usually occur following blunt high velocity trauma and often present 2–6 weeks after the injury.[1] They most commonly affect the frontal branch of the artery due to its relative exposure as it sweeps around the temporal bone and superior orbital region. Demographically young men are the most likely to present with the condition, followed by the elderly secondary to accidental falls.[2]

Whilst often easily recognised due to their pulsatile nature and presentation, care should be taken with non-pulsatile lumps of the temporal area as a thrombosed STA aneurysm may be missed. Differential diagnoses of lumps in the fronto-temporal region include

* Corresponding author. Tel.: +44 7956129103.
E-mail address: jakesloane@hotmail.com (J. Sloane).

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Fig. 1. Pre-operative marking photo demonstrating the dimensions of the lump over the patients left eye brow.

old haematoma, lipomas, neuromas, cystic lumps or parotid gland tumours. A hand held Doppler probe will usually confirm the presence of arterial flow within the lump, however if there is any doubt in the diagnosis, CT or MR angiography are the gold standard investigations.[3] Needle aspiration or core biopsy of such lumps prior to such imaging has the obvious potential to cause bleeding.

STA aneurysms can sub divided into false (pseudo) or true aneurysms and these are differentiated by histological examination after resection. False aneurysms are much more common and account for some 95% of such cases.[4] These develop either through pure mechanical shear injuries to the vessel intima or can develop more gradually due to localised necrosis of a sector of arterial wall.[5]

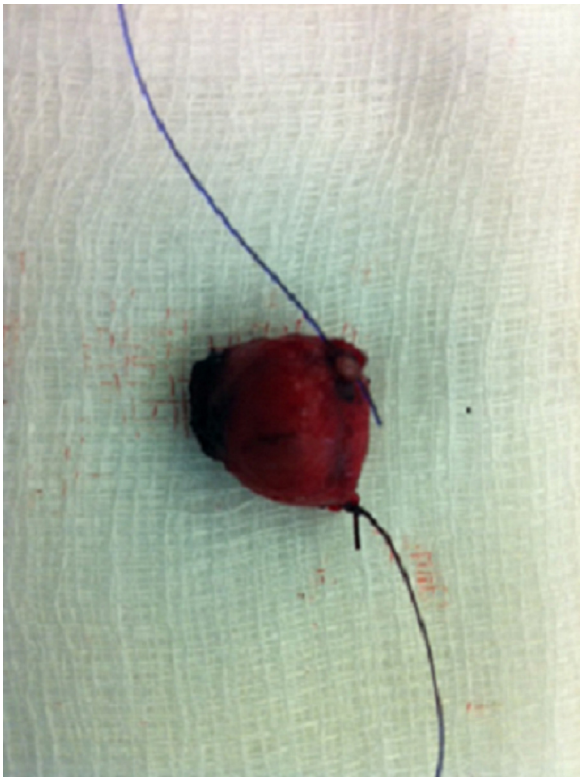

Fig. 2. The STA aneurysm fully excised with its ends ligated.

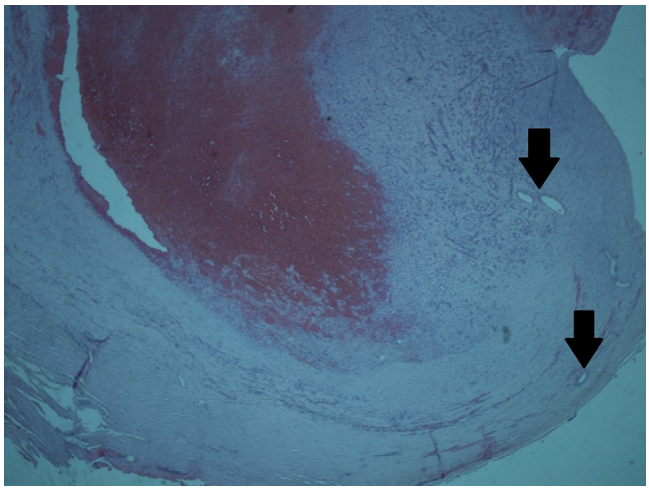

Fig. 3. Histological photograph demonstrating organised thrombus within the STA lumen and the disrupted architecture of the walls of the vessel containing myxoid/mucoid deposits. The arrows indicate several small discrete cysts noted within the adventitia.

In contrast, true aneurysms of the STA are extremely rare and are defined as involving all three layers of the vessel. To our knowledge, less than 21 cases of STA true aneurysms have been reported to date, the first being in 1954.[6]

It has therefore been proposed that true aneurysms of the STA may have a differing underlying aetiology compared to pseudo aneurysms although no firm evidence exists to prove this. Pre-existing vessel disease including congenital deformations and atherosclerosis have been theorised as possible precipitants of the condition.[5] A true aneurysm of the STA may then subsequently develop after an insult such as trauma. The association of STA aneurysms with trauma has been recognised for some time with Winslow in 1935 reporting a series of such cases secondary to duelling injuries.[6] There are however a few reports of STA true aneurysms that developed spontaneously in the absence of trauma[7,8] and demographically several of these cases include young patients who would not normally be associated with peripheral vascular disease. This may suggest a yet unknown and underlying pathology leading to an increased chance of STA aneurysm.

Our particular case was associated with further unusual histological findings. Significant myxoid and mucoid degeneration of the elastin fibres was noted. Such architectural changes represent the changed appearance of normal connective to a more gelatinous form, often post-trauma. For example such changes are well documented in meniscal tear injuries of the knee. However, such changes also found in an uncommonly seen condition – cystic medial necrosis.

Cystic medial necrosis is characterised by the fragmentation of the elastic fibres and accumulation of mucoid substance in the media of the arterial wall.[9] Defective collagen cross links with a reduction in elastic tissue of the media layer is the underlying pathology. The ineffective repair process of the artery results in a focal accumulation of ground substance, which in turn causes disruption in the continuity of laminae and muscle fibres. The weakened strength of the media increases the susceptibility of the artery to aneurysm formation.[10] In a very similar case to ours, a spontaneous aneurysm of the internal carotid artery secondary to this condition is described.

In an advanced form, myxoid changes are characteristic of Marfan's syndrome, a well known connective tissue disorder that is often associated with abdominal and thoracic aneurysms. Our subject had no other symptoms suggestive of Marfan's syndrome,

however he was contacted retrospectively to screen for both types of aortic aneurysm. No abnormality was found. In patients without Marfan's syndrome, cystic medial necrosis occurs more frequently in those of advanced age and with hypertension.[11]

The cysts noted in the adventitial layer in our case are also unusual. Atkins and Key in 1947 described a condition known as cystic adventitial disease.[12] This has come to be known by several different names, presumably due to the infrequency of its presentation. It is characterised by the deposition of gelatinous material between the media and adventitial layers. As it has been documented in childhood as well as in adults, it is not thought to be a degenerative process. Due to its similarly to the previously described cystic medial necrosis, we propose that both cystic medial necrosis and cystic adventitial disease represent a similar degenerative process with an unknown aetiology.

3. Conclusion

True-type STA aneurysm is rarely seen. The management of these includes watchful waiting, compression, embolisation, endovascular obliteration and surgical excision. Surgical management of such lumps remains the definitive method and should be considered when there is an increase in size or pain, or when the lump becomes cosmetically deforming.

Due to their rarity, it is conceivable that an underlying pathophysiology predisposes their formation. Histological analysis of the specimen in our case demonstrated features consistent with both cystic medial necrosis and cystic adventitial disease. Due to their overlap and relative obscurity, it is possible that both conditions are variants of the same underlying process. The finding of rare aneurysms and atypical histology findings should prompt the search for an underlying connective tissue disorder.

Conflict of interest

The authors declare no conflict of interest.

Funding

None.

Ethical approval

Our subject in the case was a willing participant in this case report and signed a written statement.

Author contributions

Jake Sloane – main author; Abdul Aziz – surgical assistant; Khalid Makhdoomi – lead surgeon and supervisor of writing; Samiya Ibrahim – histopathology reporting.

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